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Parents' caregiving approaches: facing a new treatment alternative in severe intractable childhood epilepsy

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Parents of children with severe and intractable epilepsy face profound caregiving challenges, dealing with their child's frequent and intense seizures, accompanying physical, social and psychological problems, and ongoing quest for seizure control through a variety of medications, diet and surgery. With the advent of a new, surgical treatment for epilepsy, vagus nerve stimulation (VNS), these parents have been presented with a renewed possibility of seizure control for their children. While many studies have investigated the effects of VNS on seizure frequency and intensity, none have looked at parents' experiences in facing this potentially life changing treatment for their child. This multiple-case study addresses this gap by exploring the experiences of nine parents of children receiving VNS. Collected over a 6-month period following parents in the hospital, clinic and in their homes, data from 22 in-depth interviews revealed that parents facing a new treatment alternative for their child experienced uncertainty around treatment efficacy and had a need to exert control over their expectations. Ongoing caregiving approaches adopted by these parents were consistent with existing literature on families living with childhood chronic illness, however, new insights were gained from parents' sharing of positive life perspectives gained through their experiences. These findings provide guidance for health care professionals working with the parents of children with severe, intractable epilepsy.

Key words: chronic illness; caregiving; epilepsy.

INTRODUCTION

Epilepsy is the most common chronic neurological condition in childhood, affecting an estimated 2.4 children per thousand[1]. Of these, between 15 and 40% will experience severe, intractable seizures, that is, seizures that continue to persist despite optimal medical treatments[1–7]. Most recently, vagus nerve stimulation (VNS), has been introduced as a surgical treatment for epilepsy. As a new treatment alternative, there has been much interest within the clinical and research communities about the effects of VNS on seizure frequency and intensity. However, to date, there have been no studies about the experience of parents facing this potentially life altering treatment for their child. Therefore, the focus of the present study was to investigate how parents of children with severe, intractable epilepsy approached VNS and the multitude of caregiving issues related to their child's condition and its treatment.

LITERATURE REVIEW

Severe intractable epilepsy in childhood

Epilepsy in childhood is a heterogeneous group of disorders representing a wide range of seizure types and frequencies. While many children with epilepsy lead relatively normal lives, a significant minority are bothered frequently by their conditions and are limited in their usual activities, and therefore, are considered to have severe conditions[1]. Most commonly, severity is considered to be a function of seizure frequency[6]. Other indicators of severity pertain to intensity, and include the occurrence of generalised convulsions, the

occurrence and frequency of seizure-related falls, injuries, incontinence or automatisms, and the length of time required to recover completely following seizure activity[8]. By extension, severity is related to seizure control. Therefore, children with intractable or difficult to treat seizures are considered to have severe epilepsy[8].

For the child with epilepsy and his or her family, severity has implications beyond the experience of seizures. Children with severe intractable epilepsy often have associated neurological impairments, developmental delays and/or psychosocial problems[6]. Effects on the developing brain such as intellectual deficits, memory and attentional disorders, and motor impairments may arise from the seizures themselves, from an underlying brain defect, or from medications used to treat the condition[4, 9–11]. Finally, the psychological and social experiences of epilepsy, including reduced independence, restrictions in activities, frequent hospitalisations, stigma, and social isolation, have the potential to limit childhood development and decrease quality of life for the child and family[12].

Seeking control: treatment alternatives in severe intractable childhood epilepsy

The goal of treatment in epilepsy is to control seizures by eliminating or minimising their frequency and intensity. For more than 60 years, the mainstay in epilepsy treatment has been the use of antiepileptic drugs (AEDs). Today, with a wide array of such medications to choose from, approximately 75% of patients achieve satisfactory seizure control with AEDs[5]. Other treatment alternatives in severe, intractable childhood epilepsy are epilepsy surgery, diet therapy and, most recently, VNS.

Vagus nerve stimulation

VNS is a new treatment for controlling intractable seizures which was introduced as an adjunct to treatment with antiepileptic medications. VNS involves the implantation of a pacemaker-like device that delivers intermittent electronic impulses to the left vagus nerve in the neck. While its mechanism of action is not well understood, VNS has been found to decrease both the frequency and the intensity of seizures, with individual results ranging from no change to complete cessation of seizures[2]. Studies of VNS in the pediatric population have demonstrated generally positive findings. In a review of the literature examining the use of VNS in children, Labar[13] cited evidence that VNS reduces seizure rates in children by 20–30% within 3 months of insertion, and by 40–50% after 18–24 months of treatment. VNS has also been found to lead to significant improvements in the child's quality of life, including improved behaviour, improved learning and social skills, reduced medication side effects, reduced postictal lethargy, increased activity, and newly acquired skills in speech, ambulation and daily living activities[14–16]. While pediatric studies are difficult to compare due to differences in methodologies, variations in seizures types, different length of follow-up times and differences in the co-administration of AEDs, they do come to similar conclusions regarding seizure-reduction and improved quality of life[13–16]. With such promising results, VNS may enable families to engage in more normal lives by improving seizure control, reducing their dependence on debilitating medications, improving their quality of life and potentially mitigating the developmental limitations their child may have experienced to date. The impact of this new treatment option on the family has not been studied.

Childhood epilepsy and the family

The few studies related to the effect of childhood epilepsy on the family report predominantly adverse effects. Findings include emotional disturbances in family members[17–22], changes in daily routines[21], social and leisure restrictions[22, 23], and economic problems associated with taking time off work for caregiving[21]. These general adverse effects might be expected to be augmented in families dealing with more severe conditions. In fact, several investigators have considered how differences in disease severity affect family members, particularly parental caregivers. Findings have shown higher levels of anxiety and/or stress in parents, particularly mothers, in families with a child with severe intractable epilepsy[19, 20, 23, 24]. In all of these studies, the greatest negative effect was seen in cases where intractability was accompanied by additional problems, such as motor and mental impairments, developmental delay and/or behavioural disturbances.

Living with a child with epilepsy: how parents manage their experience

Only a small number of descriptive studies have looked at parental approaches to managing the experience of childhood epilepsy and its effects upon the family[25–27]. Austin and McDermott[25] looked at the relationship between parental attitudes and coping behaviours in 27 parents of epileptic children, and found that positive parental attitudes towards their child's epilepsy correlated with positive parental coping patterns. They concluded that the most helpful parental responses and attitudes were maintaining

family integration, cooperation, and having an optimistic definition of the situation. Ward and Bower[27], interviewed 81 epileptic children and their parents and noted that parental confidence in managing seizures increased with their experience of seizures, and that correspondingly, they were better able to control their fears and anxieties associated with the epilepsy. Murray[26] focused specifically on parents of children with severe, intractable epilepsy ($n = 41$), and identified coping strategies that included gaining support through contact with other parents, keeping a diary, and seeking information. There have been no studies of parents' experiences managing new treatment alternatives in their ongoing attempt to achieve seizure control for their child.

In summary, children with severe intractable epilepsy and their parents face significant challenges, including frequent and intense seizures, accompanying physical, social and psychological problems, and ongoing experiences with medications, diet and surgery. Living with these challenges on a long-term basis may lead to adverse psychosocial effects on the family and parents in particular. Only limited research has been done to examine ways in which parents manage their experience with severe, intractable childhood epilepsy. While VNS offers a promising new treatment alternative, the impact of that treatment and it's associated caregiving issues have not been studied. This study attempted to address that gap in the literature by exploring how parents of children with severe intractable epilepsy managed issues around caregiving and treatment, specifically the treatment of VNS. Two research questions guided this study:

1. What are parents' expectations and responses associated with facing a new treatment option (VNS) for their child with intractable epilepsy?
2. How do parents of children with severe intractable epilepsy approach caregiving issues around their child's condition?

METHODS

Study design

A multiple-case study design was used. Case studies shed light on problems or issues as they arise in the clinical setting, and therefore, by definition, require that research questions emerge from clinical experience[28]. In addition, case studies are the preferred strategy when the research questions under investigation focus on phenomena within a real life context[29]. In the present study, the experience of undertaking a new treatment alternative (VNS) occurred within the real life context of a parent living with a child with severe intractable epilepsy, and, therefore, the case study method facilitated the identification of specific factors affecting these parents and their unique responses to them.

Sample

Selection criteria

Parents of children with severe and intractable epilepsy that received implantation of a vagus nerve stimulator at a large, metropolitan Canadian children's hospital were eligible to participate. Six children underwent the procedure during, or just prior to, the 6-month period that the study was conducted. Parents of these children were invited to participate and all agreed to take part in the study.

Child characteristics

The children, two boys and four girls, ranged in age from 3 to 14 years, with a mean age of 10 years. They had experienced seizures for most of their lives, with the age of their first seizure ranging from 1-day-old to 1.5 years old. With the exception of one child whose seizures were temporarily controlled on a nonsustainable diet therapy, the other children all experienced daily seizures, ranging from 1 to 200 per day. Each child was receiving two to five medications at the time of VNS insertion. Throughout their lifetimes, the children had been tried on between 7 and 14 antiseizure medications, alone or in various combinations, with a mean of 10.7 medications per child. Four children had tried diet therapy and none of the children had undergone epilepsy surgery.

Parent characteristics

Participants included six mothers and three fathers. Seven of nine parents were employed. Their estimated socioeconomic status varied from low (receiving welfare support) to high (double-income professionally employed homeowners). Two of the nine parents were raising their child without a partner. Two had daily professional caregiver support at home. Six parents had other, nonepileptic children at home.

Data collection

All parents were followed for a period of 6 months from the time of enrolment. Repeated contact allowed for the establishment of a trusting relationship between the parents and the investigator, providing a grounded contextual understanding of significant factors, events

and communications. Two sets of parents had children who had undergone VNS insertion just prior to beginning the study. All other parents were met prior to the VNS procedure, and were followed throughout the surgery and in the weeks and months following.

Data were collected using unstructured and semi-structured interviews with parents. The use of interviews gave parents the opportunity to express their own interpretations and understandings of their experiences. Over time, questions evolved based on data generated by the parents, and interviews became more focussed on issues of concern to a particular family, the new treatment and its effects. Data collection took place in the clinic, over the telephone, and at families' homes. A minimum of two 1-hour home visits per family were conducted, in addition to clinic visits and telephone follow-up. At least six contacts were made with one or both parents, and 22 in-depth interviews were conducted. Field notes were recorded immediately after each interview with a parent or parents. The conversations were replicated from memory as closely as possible using a verbatim account, together with contextual observations.

Data analysis

Data analysis was conducted concurrently with data collection. Themes arising from the data were conceptualised and coded into categories. An open coding technique was utilised as advocated by leaders in the field of qualitative research[30, 31], and comprised the labelling of phenomena, the successive breakdown of data into categories, and an inductive drawing of relationships and conclusions. Recurrent patterning of similar events and understandings was found, leading to a comprehensive view of parents' experiences and a defensibly valid interpretation of findings. Redundancy in themes began to be observed as data collection and analysis went on, supporting the claim of comprehensibility of findings. Finally, in an attempt to best represent the true words and experiences of parents, findings from earlier interviews were reviewed with parents and were either validated or revised. The qualitative analysis software program QSR NUD*IST (Version 4) was used to facilitate the process of data analysis.

FINDINGS

Analysis of the data collected during interactions with parents revealed three main categories of findings: facing a new treatment; ongoing approaches of parents in caring for their epileptic child; and life perspectives gained through living with a child with intractable epilepsy.

Facing a new treatment

Initial contacts with families were focused on the VNS experience and related topics pertaining to seizure control and treatment. Issues that arose for parents when facing a new treatment alternative for their child included the need to *stay on top of things* and to *balance hope with realism*.

Staying on top of things

Parents were remarkably informed about treatment alternatives. For example, at least four parents had independently gathered information about VNS several years before it was offered to their child. All parents had actively established means of acquiring new information, drawing from sources such as the internet, other parents, community organisations and health care professionals. As one mother joked, 'some people watch TV for fun—we're on the internet searching out new drugs! Its what we do with our free time'. Once VNS was implemented for their child, parents were quick to gain expertise and rapidly found themselves more familiar with it than many of the health care professionals they encountered. Having specialised knowledge about VNS that others didn't possess, parents in this group considered it necessary to 'check everything' pertaining to the treatment. The constant scrutiny led to frustration and anxiety, as expressed by one mother after uncovering a knowledge deficit in one of the professionals working with her son. She stated 'I have to tell them everything. They should know that already. You know, some parents . . . don't know to ask these things. What happens to their kids?' Even when their child showed improvement, parents were reluctant to relax their vigilance, having often justifiable fears that something would go wrong in their absence.

In spite of parents' best efforts to stay on top of things pertinent to the VNS and other treatments, they experienced difficulty in evaluating the effectiveness of treatment. While the parents of three children did perceive some benefit that they cautiously attributed to VNS—in terms of a reduction in seizures or an increase in their child's level of alertness or both—the parents of the other three children either saw no benefit or remained uncertain. A frequent phrase echoed by parents when asked about whether VNS was having any effect was 'its hard to be sure'. Or as one mother said, 'well I *think* she's better, but maybe I'm just seeing what I want to see'. The uncertainty in determining treatment efficacy was significant, as

parents could not realistically observe and count every seizure, 24 hours a day. Other factors that made it difficult to assess treatment effects included the time lag before treatments started to take effect, their child's natural cycles of increased or decreased seizure activity, their own judgement biases, hopes and fears around the treatment, and concurrent changes taking place such as environmental stressors, illnesses, other medications, and dietary changes.

Balancing hope with realism

Given the potential for improvement that VNS offered to their children, one might have expected parents to be unable to contain their hopes, yet their attitudes were surprisingly calm. Following years of seeking seizure control with other treatments, VNS was approached by parents as 'just another thing to try'. Instead of hinging all of their hopes on this new technology, they expressed a conscious effort to control their expectations, recognising that VNS might not be effective for their child. One mother explained, 'you know, every time we try something with (our daughter) we get really hopeful. We tell her its going to reduce her seizures. But that's what we told her the last time. So even though we have high hopes, we try to keep them down too'. Parents held on to a belief that even if VNS didn't work, new things would come up in the future. One mother summed up this sentiment, stating, 'there's always something more to try. You've got to think like that, or else its over, right now'.

Even when it appeared the VNS had provided relief from seizures, parents remained cautious. Their experiences with various treatments in the past had led them to believe that seizure relief might be transitory. As one mother said, 'yes, we are happy. But not too happy. It might not stay like this. We try day by day. Because, well, every time we come away with another (treatment), it works well for about 3 weeks but then it doesn't work anymore'. Thus parents' careful control over their expectations was maintained throughout the study period regardless of the perceived outcome of VNS.

The bigger picture: ongoing approaches of parents in caring for their epileptic child

In addition to concerns related to VNS, parents managed a larger group of caregiving issues on an ongoing basis. Several approaches were identified that highlighted how parents had come to deal with those issues. They included *trusting their knowledge*, *trusting others*, *promoting the best they can be*, and *proclaiming the positive*.

Trusting their knowledge

Parents possessed a deep assurance in their own knowledge about their child's state of well being and their ability to respond to events related to their child's epilepsy. Typically communicated in subjective terms, such as 'just knowing' or, 'I can tell, but other people can't', this knowledge often came across as ambiguous, inherent or instinctual. Parents specified, however, that this knowledge was not inherent at all, but rather had been acquired through years of experience. Several parents described their gradual progression from inexperience to confidence in their own knowledge and caregiving abilities.

Such confidence was often called upon when parents needed to persuade others of their knowledge, particularly when their child's response to treatment was unusual or unexpected, such as a paradoxical reaction to medication, unusual or adverse side effects, or difficult-to-observe seizures. One mother described an incident when she found it necessary to convince a clinic physician that her child was in status epilepticus, a period of continuous seizure activity constituting a medical emergency. She related:

> The doctor said to me 'what's wrong? He's fine'. And I said that he wasn't because I knew that he wasn't like he was normally, but it wasn't obvious to others. He didn't want to believe me. Anyway, they did an EEG and it showed that he was in status (epilepticus). So for a long time afterward, when I would come into the clinic, the doctor would ask me, 'how is he right now? Is he okay or not'?

Trusting others

In spite of their expertise and self-assurance, parents' access to trusted professionals and informal caregivers was extremely important to them. All parents had, over time, carefully screened and selected a small circle of individuals and organisations they felt they could count on to respond appropriately in situations pertaining to their child's care, including seizure crises. Three parents—each of whose children had gone into status epilepticus while in the care of others—reiterated their confidence in these trusted caregivers by acknowledging that these people had handled things in a manner that was consistent with the way they themselves might have approached the crisis. Having such caregivers provided parents with some respite and peace of mind, as one mother acknowledged when she said, 'even if I were there I wouldn't do anything differently'.

The strict selectivity that parents exercised over who was included in this trusted network made any change to the mix difficult and stressful. Finding an acceptable school, hospital or physician was described as a

laborious process for parents, who had experienced the need to evaluate many different alternatives for their child while at the same time meeting often inflexible eligibility requirements. One father described the intimidating process he faced in trying to get his daughter into a special school of his choice, saying 'you know, its like, there were 13 people around the table and if one of them said no then she wouldn't get in. So I knew how I had to talk to them'. Another mother, prior to her family moving to a different city, expressed her anticipation of the work involved in putting together a caregiver network, saying, 'I'll have to start again. That's why I'm going to take some time off before I start working again'. She elaborated upon her strategy of finding a specialist for her daughter, saying, 'I'm going to call this mother that (the nurse) told me about—she moved there from here recently, and she's already gone through three neurologists. She should know who's good by now'.

Promoting the best they can be

Parents of this group of children took every opportunity to promote what they deemed to be the best opportunities and environment for their child. For all parents, a primary goal was to maximise their child's ability to participate in their environment, and achieving seizure control while minimising the effects of drugs was often linked very closely to parents' goals of promoting the best their child could be. A common concern of parents was their child's level of alertness, often impaired by the cumulative effects of antiepileptic medications or postictal lethargy associated with seizures. One father expressed his frustration with the effects of medications by asserting, 'she sleeps all the time. It's no good for her. She's not living like that'. On the flip side, after achieving seizure control with the VNS and, therefore, a decrease in her child's drowsiness, one mother rejoiced that: 'she's awake, she's not sleeping all of the time. I mean, she's living'!

Some differences in the approaches of parents were noted, seemingly associated with parents' perceptions of the outcomes of treatment. The parents of two children who perceived that their child had benefited from VNS expressed their eagerness to take advantage of this respite by aggressively promoting their child's learning and development. One mother explained: 'right now, I just want to do everything for her, because she is well. I want to take advantage of the time (that she is seizure free)'. Another mom postulated: 'I think that if we could stop some of these things, if we could stop some of what's going on in her head, she might have a chance to learn'. In contrast, the parents of two children who perceived little or no benefit from the device were notably less focussed on accomplishing developmental milestones, acknowledging that their child may never achieve these milestones. One mother explained, 'at the beginning, we were always trying to push her to learn. We were always pushing. But now its better. We aren't so worried about that anymore. We had to learn to think about it in a different way. We want to make sure she can be happy, right now'.

Whatever their focus, whether it was to promote learning and development or simply to create an optimal environment for their child, every parent had found it necessary to initiate and drive changes pertaining to their child's care. Parents' efforts spanned a wide variety of causes, and included pursuing new treatments, seeking opportunities and supportive resources for learning and development, and pushing for community changes such as improved handicapped access in public places. For example, motivated by their own children's circumstances, the parents of the first two children to receive the VNS at the study hospital were the driving force behind generating enough public awareness and acquiring the necessary funding to allow a total of 16 children to receive the previously unfunded procedure.

Leading such changes often required an immense amount of energy, resourcefulness and persistence, and sometimes parents questioned their endurance. Coming up against what at times felt like continual obstacles, one mother described being tired of 'fighting' for things all the time. Other parents also experienced fatigue associated with pushing changes through, but at the same time they took pride in their ability to make things happen. This sentiment was summed up by one mother, who stated, 'they have seen me so much over the years and they know I will argue until I get what I want. When its for (my son) I don't mind pushing'.

Proclaiming the positive

Parents expressed a deep appreciation of their children, and every parent interviewed was eager to communicate the good or special qualities of their son or daughter. After being invited to describe her son, one mother questioned whether she was being asked to talk about his 'problems', and was visibly surprised and pleased at being encouraged to share how 'wonderful' he was. Parents described how smart their child was, how courageous, how beautiful, and how good natured they were. Often this was reinforced by contrasting their child favourably with other children who were chronically impaired. Other times, parents communicated their feelings of pride by emphasising how much other people appreciated their child. This sentiment was summed up by one mom, who said of her son, 'oh, they all love him. Everybody loves him'!

Life perspectives gained through living with a child with intractable epilepsy

From parents' descriptions of their daily challenges and responses, three themes emerged that seemed to reflect their pervasive emotional perspectives rather than their active approaches to caregiving. The theme, *being scared*, reflected parents' experience of living with fears around their child's condition. The themes, *living 'day by day' and 'I do what I have to do'*, represented fundamental shifts or philosophies in how they approached and lived their lives.

Being scared: fear of future losses

In the midst of their remarkable calmness in the face of seizures, new treatments and surgery, parents talked about being scared. For the most part, such fears were not readily apparent in daily interactions, but they were close to the surface and could be triggered by events or conversations. Each parent described a different fear, however, they all seemed to be associated with the possibility of future loss in some form: either loss of their child's life, a decline in their child's level of functioning, or loss of hopes for their child's future. Several parents spoke of their fears of adverse outcomes resulting from seizures as the only reason they were prepared to keep trying new treatments. As one mother said, 'what really scared me was when they told me what a risk it was when she was having the seizures. They said her heart could stop. After that, I was ready to try anything. Just bring it on'. Another mother spoke of her fear of running out of treatment alternatives. She commented, 'you know what I'm really scared about? I'm scared of the day when there's nothing left to try. That really scares me. Because I'm not ready to give him up yet'. Other parents expressed fears associated with their child's future. As one mother described, 'what scares me is what will be the outcome of her life. Will she be independent? Will she have a life of her own? What will she be able to do? I don't know these things, so that's what I'm always afraid of'. Whatever the particular focus of their fears, living with fear was an ever present condition of these parents' lives.

I do what I have to do

The parents all managed to do what was necessary to care for their child with severe intractable epilepsy. In practical terms, different parents managed their situations in entirely different ways. At least four parents had made specific career changes based on their child's caregiving needs, yet in spite of such changes, most parents were actively involved in work outside of the home and were advancing their careers. Two parents had separated from spouses after their children had developed epilepsy and were raising their children alone, yet these were the same two parents who had acquired daily caregiver support at home. Some parents experienced reduced social lives while others were very active socially. The common denominator seemed not to be the specific lifestyle accommodation, but rather their unquestioned acceptance that their lives had changed and that they would do whatever they needed to do to care for their child. Parents expressed exhaustion, frustration and impatience, yet they never expressed resentment or unwillingness to do what was necessary. A powerful example of the strength of this acceptance was seen in how much one mother was prepared to forfeit, when her husband questioned the sacrifices they were making as a family:

> He wanted to put her in another home where they would take care of her and she wouldn't live at home. He didn't want her to live at home. He thought it was too hard. So I told him if he needed a rest, then he could take one. I would stay with (our daughter). So he went away for a week, and then he came back.

Living 'day by day'

Several parents remarked on what they considered to be a positive change in their lifestyle as a result of having a child with severe, intractable epilepsy. Parents commented upon how formerly, they had been more prone to plan ahead, to worry about the future, and to try to control events. Now, several parents remarked, they had adopted a 'day by day' approach. Living with multiple uncertainties, including seizure occurrence, treatment efficacy, or their child's developmental progress, they no longer attempted to predict or control outcomes in the way they did before. As one father stated, 'we have learned patience. Because (our son) doesn't do things at the same time as other children. It takes a long time for him to develop. So we have to be patient. So we take life day to day'.

Rather than viewing their limited planning horizon with frustration, parents recognised it as a positive change in their thinking and something that they valued. Parents' acknowledged how the hardships of living with chronic epilepsy had enhanced their everyday outlook on life. The mother of a 3-year-old boy said, 'I have learned to appreciate life more . . . I appreciate what (my son) has. We are so happy every time he does something new. I think we are even more happy than we would have been. I think it has made me not worry about the small things'.

DISCUSSION

The findings of the present study illuminate a set of perspectives, approaches and strengths that enable parents of children with severe, intractable epilepsy to manage their significant caregiving challenges. While a number of findings are consistent with the more general literature on uncertainty in chronic illness and families living with childhood chronic illness, some are unique to this study and provide new insights into parental experiences in caring for a child with severe, intractable epilepsy.

Parental limbo: uncertainty versus control

The issues raised by parents facing the new treatment of VNS are consistent with well developed concepts of uncertainty and control in chronic illness. Much of the stress families face in having a young member with a chronic condition has been attributed to uncertainty[32–34]. Parents of a chronically ill child live in what has been described as a state of 'sustained uncertainty' an unremitting experience of uncertainty from which they cannot achieve respite[32]. Uncertainty is associated with a lack of knowledge, predictability and control, and correspondingly, reported strategies to relieve uncertainty have been those that help parents manage information and thereby regain a sense of control[25, 32, 33, 35, 36].

Uncertainty has been reported as being especially pervasive for parents of children with epilepsy[25, 26, 37]. Murray[26], in particular, elaborated upon the uncertainty faced by parents of children with severe intractable epilepsy and described five key dimensions of parental uncertainty. These dimensions were associated with the child's diagnosis, etiology, seizure activity, treatment and prognosis. In the present study, parents' frequent comments that 'its hard to be sure' were an expression of their uncertainties around the occurrence of seizures and the efficacy of treatment. Similar to Murray's findings, uncertainty for parents was exacerbated by the characteristic unpredictability of seizures, changes in the disease and in treatments, the ever-looming possibility of seizure control, and uncertainty about future outcomes for their child. Parents' attempts to alleviate these uncertainties were observed in their efforts to stay informed about treatments and to verify information to ensure the safety and well being of their child.

The phenomenon of parents' purposeful attempts to balance or constrain their hopes for achieving seizure control also fits well with the central concepts of uncertainty and control. Although it has not been previously described in the epilepsy or chronic childhood illness literature, this finding can be conceptualised as a response to help control the uncertainties associated with the new treatment and its outcome. For years, each new treatment including VNS, had brought the promise of improved seizure control. The intermittent renewal of hopes surrounding each new treatment had forced parents into a roller coaster of raised expectations and subsequent disappointments. Living in this kind of limbo, parents attempted to concurrently sustain yet constrain their hopes and expectations, thus protecting themselves emotionally from uncertain outcomes.

Ongoing approaches to caregiving

The ongoing caregiving approaches of parents in this study echo much of what has been reported in the literature pertaining to parental experiences in caring for a chronically ill child. Several investigators have observed that with exposure to the challenges of chronic illness over time, families develop strategies, skills and resources that empower them to manage their situations more effectively[25, 35, 38–40]. In particular, several studies of the approaches of parents of chronically ill children described a 'taking charge' phenomenon in which parents had learned ways to participate actively in planning care and advocating for their children within the health and educational systems[35, 39, 40]. These previously documented examples of parental leadership correspond to observed strategies of parents in the present study, particularly their ability to stay informed and utilise their expertise, actively promote the best conditions for their child, and establish a trusted support network.

Positive outcomes and changed life perspectives

To date, much of the research literature on the impact of severe intractable epilepsy on parental caregivers has reported negative outcomes, such as anxiety, stress, lifestyle restrictions and economic burdens[17–23]. Consistent with such findings, parents in this study experienced high levels of anxiety, stress and lifestyle changes. It seems probable that the intense demands placed upon them in terms of expertise, vigilance and leadership were far greater than those required of parents of unaffected children, and that these strengths came at a cost. Their frequent expressions of fatigue, their inability to relax their vigilance, and their pervasive state of fear for their child and for their child's future all lend weight to this hypothesis. One might even speculate that these parents were so involved with their child with severe, intractable epilepsy, that other parts of their lives,

their careers, their marriages, or their relationships with their unaffected children had been negatively impacted. However, while parents in this study found it necessary to make a variety of life adjustments, there was no conclusive evidence pointing to specific detrimental effects. In fact, what stood out was parents' determination to assert the positive outcomes they had experienced.

Evidence for positive outcomes is seldom seen in the epilepsy literature. Within this study, however, there were several indicators that parents' experience of living with and caring for a child with severe, intractable epilepsy provided them with positive insights and strengths that seemed to have occurred concurrently with their hardships. For example, parents' efforts to promote the best conditions for their children resulted in remarkable demonstrations of initiative and leadership that often resulted in benefits to others. In addition, parents' day by day approach to life came across, not as a sacrifice, but rather as a positive, valued change in their philosophy of living that allowed them to appreciate life to a fuller degree. While they were limited in their ability to plan for the future, they viewed their day to day approach to life as one that allowed them respite from worrying about superficial matters or unalterable contingencies. Finally, parents' voicing of their child's unique and wonderful qualities came across as a desire to communicate that, in spite of their child's deficits, he or she was special and valued. Ultimately, this 'need to tell' seemed to stem from their deep appreciation of their child and of what their child had brought to their lives.

Implications for practice

These findings have direct implications for health care professionals working with parents of children with severe, intractable epilepsy. Acknowledging parents' expertise is a critical first step for the clinician in order to foster the mutual exchange of information and gain the most accurate and complete clinical picture. As these parents have emphasised, it is important that health care professionals be accessible, listen openly and give credence to their concerns. Clinicians can then help to disseminate parental knowledge and expertise to other caregivers, and can act as an additional resource to supply information and support.

This study draws attention to the fact that different parents will be at different places in terms of their priorities and their support needs, and, therefore, the need for thorough and in-depth family assessments becomes key. Understanding the trajectory that parents may follow, clinicians can provide validation to less experienced parents to help build their confidence in their own knowledge and skills, and thus can teach them how to advocate for their child. For more experienced parents, such as those participating in this study, clinicians can be attentive to the differing priorities of parents in promoting the best that their child can be and help them to find the resources to achieve this. Finally, this study underscores that parents at all levels of experience and expertise become exhausted and overwhelmed by the incredible challenges they face in caring for a child with severe, intractable epilepsy, and thereby highlights the need for adequate respite care.

Limitations of the study

In this study, attempts were made to include a diverse group of parents and children, however the sample size was small and, therefore, data may not accurately represent the views and approaches of a larger group of parents of children with severe, intractable epilepsy. In addition, parents who were more involved in caregiving contributed a proportionately greater amount of data to this study. Six mothers and three fathers participated in the study, and therefore findings may represent a bias towards maternal approaches and perspectives.

Future research

Further research needs to be done into the experiences, approaches and perspectives of parents of children with severe, intractable epilepsy, beginning with a replication of this study with a larger group of parents to validate current findings. In particular, the phenomenon of changed life perspectives—a finding that emerged spontaneously as a result of extended contact with parents—has not been seen in the research literature to date. Further exploration of this finding may help to gain a richer and more complete understanding of how parents' perspectives have shifted as a result of their unique and challenging caregiving experiences.

CONCLUSION

This study set out to explore the expectations and approaches of parents of children with severe, intractable epilepsy as they faced a new treatment alternative, VNS, and as they engaged in their everyday caregiving activities. The findings revealed that these parents had developed a unique set of strengths and life perspectives that allowed them to manage their significant caregiving challenges. As an important part of these parents' valued support network, health care professionals are in an ideal position to work

with, validate and support these parents as they care for their children.

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